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Infiltrative, ulcerative, and fistular lesions of the penis due to lymphogranuloma venereum

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In some parts of the tropics lymphogranuloma venereum (lymphogranuloma inguinale) can still be regarded as an important venereal disease. In the past, between 1925 and 1940, it was a dreaded disease in Europe. In those years the disease had for some unexplained reason also become endemic in Finland, affecting about 2,000 people, 500 of whom were females. The serious, sometimes fatal, late manifestations involving the female genitalia and rectum made even syphilis appear to be mild in comparison. Contrary to the high percentage of late complications in females, the late manifestations in males (homosexuals apart) were fairly rare.

Elephantiasis of the penis and scrotum is probably the best known of the male complications. As to the pathogenesis of the condition, we have to consider not only obstruction of lymphatic flow caused by involvement of the inguinal and iliac lymph nodes, but also the possibility of an active diffuse lymphogranulomatous infection in the subcutaneous tissue.

May and Castiglioni (1938) listed Peyronie's disease among the lymphogranulomatous complications. In the few cases of this condition that we have seen, no connection with lymphogranuloma could be found and we have consequently excluded Peyronie's disease from our review of the literature.

In the present paper we consider only those cases of lymphogranuloma venereum in which the infection had led to the development of preputial infiltrates, penile lymphangitis and bubonuli, with or without fistulae, and more or less chronic ulcerative processes. Altogether 37 cases of this kind have been observed in Finland. Data relating to the clinical course and laboratory findings in four of them (Cases 1–4) were published by Cedercreutz (1934a, b). In this report we present a survey of all 37 cases and, in addition, eight case histories which illustrate the severity of these rare conditions.

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Review of the literature

PENILE LYMPHANGITIS AND BUBONULI

In some cases of subacute lymphogranuloma venereum in males, as well as the inguinal lymph nodes the lymphatic pathways from the initial site of infection may be involved, particularly along the dorsum of the penis, and may show lymphangitis. Ravaut, Boulin, and Rabeau (1924), in France, were among the first to pay attention to this lymphangitis and bubonulus formation on the penis.

Ravaut and Scheikevitch (1921) had reported 3 years earlier a preputial nodule, the size of a nut, which showed a fistula into the skin. Similar infiltrates in the early stage of lymphogranuloma venereum were described by other workers, mostly as a nodular type of primary lesion, or as a *chancre en noisette* (Sézary and Facquet, 1934; Sézary and Drain, 1935; Weissenbach and Témime, 1939). However, according to Cedercreutz (1934a, b), this type of lesion is not a primary lesion but a small bubonulus with fistula formation. Cases of penile lymphangitis or abscess formation and fistulae were further reported from France by Lévy-Franckel and Temerson (1934) and by Weissenbach, Boccage, and Bouwens (1938).

Frei (1932), who introduced his skin test in Germany in 1925, estimated the frequency of lymphangitis to be about 4 to 5 per cent. of his cases. Further cases were reported from Germany (Buschke, Boas, and von Vásárhelyi, 1931; Froboese, 1933; Löhe and Rosenfeld, 1932), from Switzerland (Burckhardt, 1934), from Italy (Midana, 1938; Peruccio, 1937; Tarantelli, 1935), and from Spain (de Gregorio, 1937a, 1944). From the pus of a suppurating bubonulus formation, which had developed in connection with a dorsal lymphangitis on the penis, de Gregorio was able to prepare a Frei antigen which produced strongly positive skin tests in lymphogranuloma patients. In a Rumanian series

(Nicolau and Banciu, 1932a, b), lymphangitis of the penis was seen in 2·6 per cent. of cases. One of a series of experimental inoculations of lymphogranuloma agent into patients suffering from general paralysis resulted in a penile lymphangitis also (Wassén, 1933).

Only occasional cases have been reported in the North American literature (Sheldon and Heyman, 1947; Smith and Custer, 1950), but there have been numerous reports from South America, as can be seen in the large monograph by May (1940). Bubonuli were reported by Bonnecarrère (1939), Castiglioni (1938), May (1940), and Quiroga (1937, 1938); and dorsal lymphangitis by Coutts (1935, 1938), Coutts, Vargas Molinari, and Lecaros Matte (1938), May and Castiglioni (1938), Riveiro Rivera (1938), Rovira Burzaco (1939), and Agneta (1948).

According to May (1940), lymphangitis is fairly characteristic in lymphogranuloma inguinale. Many cases of lymphangitis described earlier in connection with either syphilis or gonorrhoea, may in fact, have been due to lymphogranuloma.

ULCERATIVE LESIONS ON THE PENIS

The first cases of chronic ulcer of the penis due to lymphogranuloma (ulcus chronicum penis) were described by Nicolau and Banciu (1932a) from Rumania and by Cedercreutz (1934a,b) from Finland. The patient reported by Nicolau and Banciu had balano-preputial ulceration, 4 × 2 cm. in size, of 8 months' duration, which showed no tendency to heal. Two of Cedercreutz's cases were impressive examples of severe chronic ulceration, with involvement of the urethral wall which resulted in urethral fistulae.

Nicolau (1934) also described a case of chronic ulceration which led to a similar complication. An extract obtained from the surface of the ulcer had the antigenic properties of a Frei antigen.

Cedercreutz as well as Nicolau found it quite logical to regard these ulcerative processes of the genital organs in males as analogous with chronic vulval ulceration (ulcus chronicum vulvae) in females, in other words as *esthiomène masculinum*.

TABLE *Findings in 37 cases*

Case No.	*Age (yrs)*	*Year disease contracted*	*Primary lesion*	*Inguinal bubo*	*Frei test*	*Duration of disease*
1	33	1930	−	Left	+++	6mths
2	36	1929	+	Bilateral	++	>12 mths
3	29	1931	+	?	++	>18 mths
4	26	1931	+	Bilateral	No data	4 yrs
5	19	1919	+	Right	+++	2 yrs
6	29	1926	+	Bilateral	++	Not known
7	22	1929	+	Bilateral	++	>9 mths
8	24	1929	+	Left	Pos.	>8 mths
9	32	1930	+	Bilateral	No data	Not known
10	23	1930	?	Bilateral	++	>4 mths
11	26	1930	+	Bilateral	No data	Not known
12	25	1930	+	Bilateral	+	>6 mths
13	25	1931	−	Left	+	>3 mths
14	24	1932	+	Bilateral	+++	7 yrs
15	21	1933	+	Left	Pos.	>4 mths
16	38	1933	+	Bilateral	Pos.	Several mths
17	36	1934	+	Left	++	6 mths
18	26	1934	+	Bilateral	+	>6 mths
19	32	1934	+	Bilateral	No data	Not known
20	23	1934	+	Left	++	4 yrs
21	31	1935	+	Bilateral	No data	Not known
22	29	1936	+	Bilateral	+	10 mths
23	20	1935	+	Left	Pos.	>4 mths
24	39	1935	−	Bilateral	Pos.	>4 mths
25	17	1935	+	?	++	8 mths
26	33	1935	+	Left	±	>3 mths
27	48	1936	+	Right	+	3 mths
28	45	1936	+	Left	+	5 mths
29	25	1937	−	Bilateral	+	Not known
30	28	1937	+	Bilateral	+	>5 mths
31	27	1937	+	Right	+	Not known
32	25	1937	+	Right	+++	4 mths
33	26	1937	+	Right	++	2 mths
34	67	1937	−	Left	+++	9 mths
35	36	1937	−	Right	+++	8 mths
36	28	1938	+	Bilateral	++	>6 mths
37	32	1938	+	Bilateral	++	8 mths

According to Nicolau, the chronic ulcer could, in many instances, in males as well as in females, develop directly from the primary lesion which—contrary to its usual habit of quick healing—persisted and slowly progressed to form a large chronic ulcer. Cedercreutz, however, believed that the ulcus chronicum developed from preputial infiltrates or small bubonuli of lymphangatic origin. Two of his cases were reported to illustrate this concept. Instead of typical ulcerations the patients presented only small preputial bubonuli or lymphangitic abscess formations reminiscent of the *chancre en noisette* of the French authors. These could be regarded as forerunners of *esthiomène masculinum* in more severe infections. They are rather early manifestations in the course of lymphogranuloma venereum, mostly coincident with the development of the inguinal buboes (Cedercreutz, 1934a,b; Chevallier, 1936).

At a French symposium on chronic ulcers of the vulva in 1936, Chevallier remarked, in his leading paper, that chronic ulcers of the penis due to lymphogranuloma venereum are very rare. He had seen it develop in one case after an unsuccessful attempt to remove a bubonulus, probably not radically enough. In two other cases in which he had ventured to suggest this diagnosis, the first proved to be a cancer and the sceond a case of syphilis.

De Gregorio (1932, 1936, 1937b, 1944) described four cases of chronic penile ulcer due to lymphogranuloma venereum, two of which were conjugal cases. One of these is particularly interesting as it illustrates the long-standing infectivity of the chronic lesions. A male patient had bilateral suppurative buboes in both groins in 1920 and subsequently a chronic ulcer in the coronal sulcus. When he married some 13 years later he transmitted the infection to his wife, who in 1933 developed bilateral inguinal buboes and proctitis.

Further cases of chronic ulcerative lesions on the penis have been reported by Chauvin and Vigne (1938), Fariñas Guevara (1941), Glaze (1937), Howard, Eisenman, and Strauss (1939), Peruccio (1937), Radaeli (1938), Rechter (1943), Sărăteanu (1937), and Semmola (1939, 1940a,b). Tissue extracts from several of these ulcers proved to have the antigenic properties of Frei antigen.

In a survey of lymphogranuloma venereum

Wassermann reaction	*Year syphilis contracted*	*Penile complications*
Neg.	——	Preputial infiltrate → ulceration and sinus formation
Neg.	1921	Preputial infiltrate, size of thumb-tip. Excised
Neg.	——	Lymphangitis and penile bubonulus → large ulcer and urethral fistula
Neg.	——	Extensive ulceration and urethral fistula. Rectal stricture
Neg.	——	Bubonulus of penis. Extensive destructive ulceration
Neg.	——	Large ulcer with destruction of prepuce
Neg.	——	Preputial bubonulus (*chancre en noisette*)
Neg.	——	Lymphangitis. Odema and ulceration of penis
Neg.	1928	Preputial infiltrates. Phimosis. Dorsal incision
Neg.	——	Ulcerative balanitis. Preputial infiltrate. Excised
Neg.	1927	Bubonulus of penis
Neg.	——	Preputial infiltrate (*chancre en noisette*). Erosive balanitis and phimosis
Neg.	——	Bubonulus of penis, pea-sized
Neg.	1932	Chronic penile fistulae. Excised
Neg.	1933	Bubonulus, pea sized. Large ulcer
Not recorded	1927	Bubonulus of penis
Pos.	1934	Preputial infiltrate
Neg.	1933	Two penile bubonuli. Recurring ulceration
Neg.	1934	Periurethral abscess. Chronic ulcer (*esthiomène masculinum*)
Pos.	1934	Chronic penile ulcer. Deep fistulae
Neg.	——	*Esthiomène* and lymphangitis of penis
Pos.	1936	Bubonulus of penis. Phimosis
Neg.	——	Two small pea-sized infiltrates. Chronic ulcer
Neg.	——	Lymphangitis and perilymphangitis of dorsum of penis. Small preputial infiltrate
Pos.	1935	Chronic ulcer, resistant to anti-syphilitic treatment
Neg.	——	Preputial infiltrate
Neg.	——	Large preputial infiltrate with suppuration
Neg.	——	Preputial infiltrate → abscess and chronic ulcer
Neg.	1936	Lymphangitis of penis
Neg.	——	Bubonulus and lymphangitis of penis
Weak pos.	1937	Nodular lymphangitis of dorsum of penis
Pos.	1937	Chronic ulcer
Neg.	——	Preputial infiltrate. Excised
Neg.	——	Bubonulus and lymphangitis of penis → fistula. Excised
Neg.	——	Bubonulus of penis → chronic ulcer. Excised
Neg.	——	Two preputial infiltrates → chronic ulcer. Excised
Neg.	——	Lymphangitis with fistula formation. Excised

(8,731 males and 990 females) in India by Rajam and Rangiah (1955), the occurrence of penile ulcers and fistulae is briefly mentioned, but the exact number of cases is not given. They include illustrations of localized elephantiasis of the prepuce with discharging sinuses, chronic ulcers of the penis and scrotum, and multiple bubonuli.

CHRONIC PENILE FISTULAE

In some exceptional cases of lymphogranuloma venereum deep fistulae may develop on the penis and persist for years. Some preputial fistulae and sinuses from bubonuli of shorter duration have been mentioned above, as have also urethral fistulae (Cedercreutz, 1934a, b; Nicolau, 1934) in connection with chronic destructive ulcerations. The first report of a case of urethral fistula due to lymphogranuloma venereum was, however, presented by Kleeberg (1932). The patient, a man aged 35, had a bubo in 1920. In January, 1931, he was found to have a urethral stricture and discharge (which showed the antigenic properties of Frei antigen); the condition was complicated by a periurethral abscess and two urethral fistulae.

Severe cases of urethral, penile, and scrotal or urethral and perineal fistulae were also reported by de Gregorio and Zatorre (1949) and by de Faria (1949).

Midana (1938) described an interesting case of a deep fistula extending from the preputial limbus to the root of the penis. It had developed from a nodular lymphangitis on the dorsum of the penis. The discharge from the fistula proved to have the antigenic properties of a Frei antigen.

Present series

Adequate hospital records were available for 810 of the male patients with lymphogranuloma venereum who were seen in Finland between 1925–1940. Among these patients were 37 who showed penile complications, such as lymphangitis, bubonuli, preputial infiltrates, discharging sinuses, and fistulae.

The findings in the 37 cases (Table: pp. 194–195) include the age of each patient and the year in which the lymphogranuloma infection was contracted. The presence or absence of a primary lesion at the time of the patient's first visit is indicated by a + or − sign. The Frei test was positive in 32 cases, and was either not performed or not recorded in five. In five of the Frei-positive cases (in the Table marked 'pos.'), no details were given on the strength of the reaction. The Wassermann reaction results shown are those recorded when the lymphogranuloma infection was diagnosed. For those who had had syphilis, the year when it was acquired is given in the adjacent column.

Illustrative case reports

Case 5, a 19-year-old dealer from Kiuruvesi, developed both a persistent ulcer on the penis and a suppurative bubo in the right groin in December, 1919. The ulcer enlarged and eventually involved large areas of the penis. An abscess, probably a bubonulus, developed dorsally near the base of the penis and left a deep cavity. The patient was admitted to another hospital on several occasions, and then to the Dermatological Ward, University of Helsinki, between August and December, 1920. The Wassermann reaction (WR) and Kahn test were negative. The ulcer healed very slowly, leaving behind a scar which occupied a large part of the dorsum of the penis. Because of this scar the penis stayed in a horizontal position except at erection when it was bent upwards in a curve against the abdominal wall like a Turkish sabre. This proved to be no disadvantage; on the contrary, both his wife and himself were well satisfied with the condition.

When he was re-examined in 1938, the penis showed more scarred than normal skin, the whole anterior surface being occupied by scar tissue (Fig. 1) which in some areas was 10 mm. thick. The Frei test with two different antigens was strongly positive.

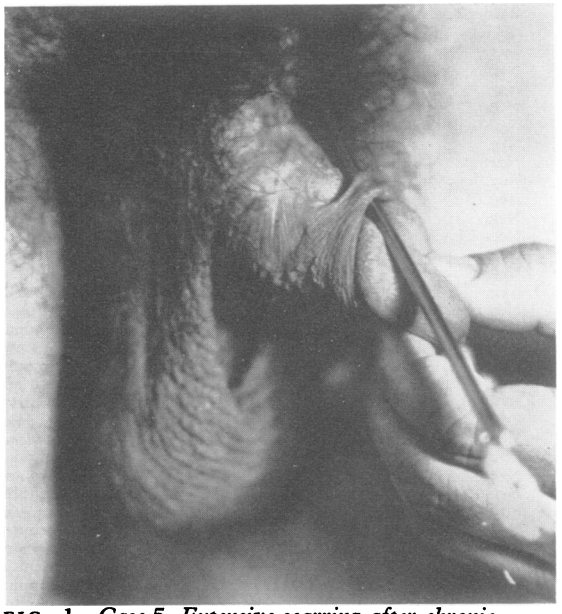

FIG. 1 *Case 5. Extensive scarring after chronic ulcer of penis*

Case 6, a man aged 29 from Suojärvi, after coitus with a prostitute in Viipuri in 1926, developed bilateral inguinal buboes, with multiple discharge sinuses. The

condition persisted for several months. He transmitted the infection to his wife, who then also suffered from suppurating bilateral inguinal buboes and later developed elephantiasis and chronic ulceration of the vulva and a severe rectal stricture. In 1930, infiltrations and ulcers appeared on the penis, with abscess formation in the posterior part of the prepuce. The frenulum and large parts of the prepuce were destroyed by the ulcerative process. Some ulcers were also seen more proximally.

When he was examined in the Dermatological Clinic, University of Helsinki, in March 1938, the patient showed extensive scar formation near the coronal sulcus and in the preputial region. The only remains of the prepuce was a tongue-like flap on the left side (Fig. 2).

FIG. 2 *Case* 6. *Destruction of the prepuce and scarring after chronic ulcer of penis*

There were some smaller scars on the penile shaft. The Frei test was positive with four different antigens. The WR, Kahn and Ito-Reenstierna tests were negative. His wife (Sonck, 1941, Case 103) also had a positive Frei test.

Case 9, a taxi-driver aged 32, from Helsinki, contracted both syphilis and gonorrhoea in December, 1928. He indulged in intercourse with many prostitutes in Helsinki, and on March 5, 1930, developed a tiny lesion on the preputial margin, followed by an inflammatory swelling of the whole prepuce, with phimosis. The inguinal lymph nodes were enlarged. 10 days later he underwent a dorsal incision of the phimotic prepuce.

He was admitted to the Dermatological Ward, University of Helsinki, from March 24 to April 14, 1930, when the diagnosis was lymphogranuloma inguinale and treated syphilis. The WR was negative. There were large strumous buboes in both groins. The distal end of the penis was very swollen in the shape of a pear and the glans penis appeared to be partly inbedded in and surrounded by preputial infiltrates (Fig. 3). Pus was aspirated from the inguinal buboes six times between March 27 and April 9.

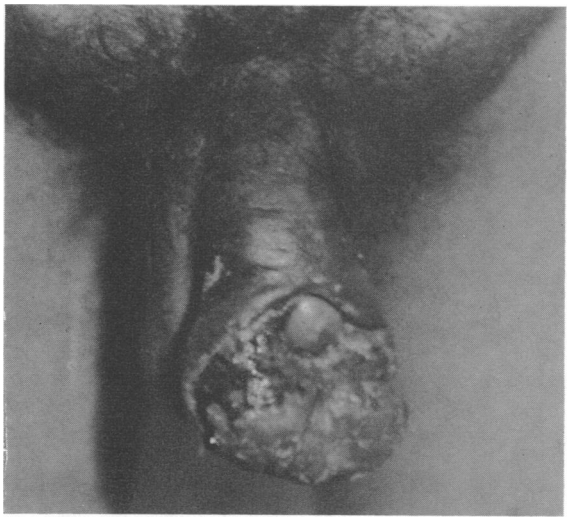

FIG. 3 *Case* 9. *Bilateral inguinal buboes. Glans penis imbedded in preputial infiltrates*

Case 14, a dealer aged 24, from Helsinki, (Sonck 1941, Case 134), had gonorrhoea on June 23, 1932, and on July 6 an inflammatory phimosis with two small darkfield-negative ulcers behind the coronal sulcus. On July 12, there were multiple ulcers and chancroid was diagnosed. On July 22, bilateral inguinal buboes were noted. These were incised on July 30 and August 8. On August 16 he had an indurated ulcer at the urethral orifice. This was darkfield-positive and primary syphilis was diagnosed. He was treated with bismuth and neosalvarsan.

He had frequent intercourse in December, 1932, and in January, 1933, he developed a preputial infiltrate, the size of a finger-tip, with two draining sinuses into the inner prepuce immediately behind the coronal sulcus. There were also bilateral inguinal buboes. The Frei and Ito-Reenstierna tests were both positive. He also had florid gonorrhoea.

Two of his contacts were found to have gonorrhoea, syphilis, and lymphogranuloma.

He was admitted to the Dermatological Ward, University of Helsinki, from April 11 to May 12, 1938, with a diagnosis of lymphogranuloma venereum, and it was found that he also had gonorrhoea. The WR and Kahn test were negative. There were several scars in the groins. The penis was swollen, with two fistulae (7 and 4 cm. long) on the dorsum of the penis and some scars more laterally. Along the dorsal midline an infiltrated cord was palpable from the coronal sulcus to the base of the penis, disappearing behind the angle of the pubis. It consisted of a fistular canal which discharged pus, and was 8 cm. long from its opening in the coronal sulcus (Fig. 4). In April, 1938, and March, 1939, Frei tests were strongly positive with two different antigens. The fistulae and infiltrates were radically extirpated in April, 1939, with subsequent satisfactory healing.

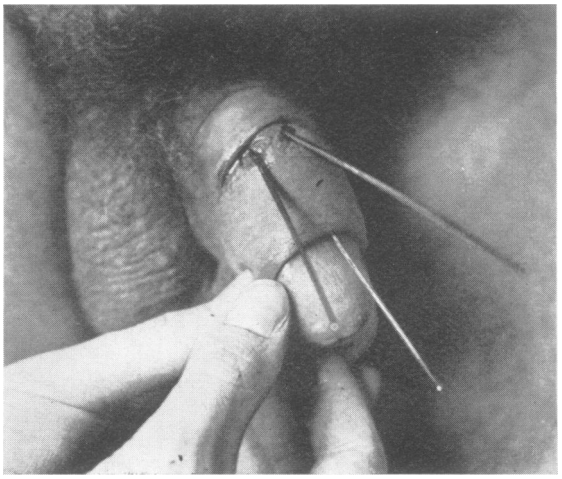

FIG. 4 *Case* 14. *Contagious lymphogranulomatous fistulae of* 6 *years' duration* (4, 7, *and* 8 *cm. long*)

Case 20, an engineer aged 26, from Helsinki, acquired simultaneously three venereal diseases, gonorrhoea, syphilis, and lymphogranuloma venereum, after his first ever intercourse in July 1934. About 2 weeks after the onset of the gonorrhoea, two small split-like ulcers appeared in the coronary sulcus on either side of the frenulum and the left inguinal lymph nodes became enlarged. Serological tests for syphilis were positive, and he was given two series of combined antiluetic treatment (20 bismuth and 16 neosalvarsan injections in all); in spite of the insufficient treatment, the tests became negative during the following 2 years.

Symptoms of lymphogranuloma venereum became obvious in August, 1934, and a suppurating bubo in the left groin was incised on August 24. Infiltrated lymphangitic cords developed on the lower side of the penis (uninfluenced by the antiluetic treatment), extending from the ulcers near the frenulum to a hard preputial infiltrate, the size of a thumb-tip. These infiltrates developed into a fluctuating bubonulus which then discharged pus through two draining sinuses at the base of the prepuce on the lower side of the penis. The skin between the sinuses became necrotic and a chronic ulcer, 3 cm. in diameter, which showed no tendency to heal, soon developed. The patient complained of intermittent severe pains in the penis which interfered with sleep. New foci of suppuration developed in the inguinal bubo. Some of these were incised, others drained spontaneously. The penile ulcer was treated with hot-water baths, wet dressings, ultra violet light, etc., and was partly epithelialized 1 year later.

In January, 1936, there was a severe exacerbation. The penis was swollen with infiltrates extending from the former ulcer in a proximal direction, and with two new ulcers leading to sinuses. The WR and Kahn test were negative, the Frei test was positive. In April, 1936, the depth of the right sinus was 3 cm. and that of the left 2·5 cm. A new ulcer developed at the base of the penis. When he was seen in August, 1936, the penis was swollen and ulcerated, and the patient's general condition poor. Intercourse was impossible. In May, 1937, for no apparent reason, there was a further severe exacerbation. Within 7 days three ulcers developed on the anterior surface of the penis and the original ulcer on the posterior surface grew larger (Fig. 5). It measured 8 cm. from right to left and was connected *via* a deep fistula with an ulcer near the base of the penis. Simultaneously with this exacerbation the inguinal lymph nodes again showed enlargement and tenderness. Persistent pains in the penis kept the patient awake at night.

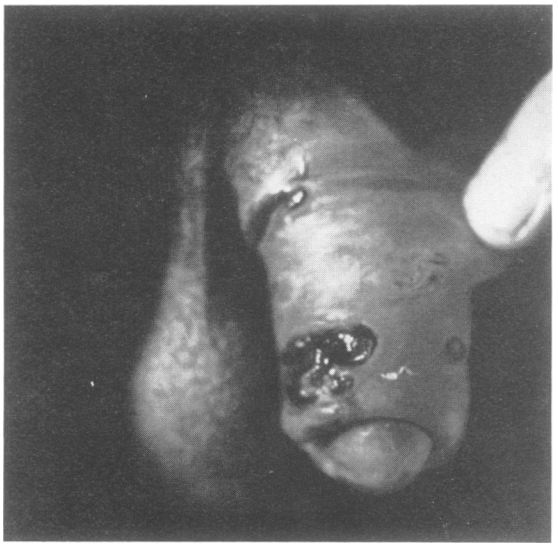

FIG. 5 *Case* 20. *Painful ulcers on penis of* 3 *years' duration* (*May*, 1937)

In June, 1937, the WR and Kahn test were positive. The erythrocyte sedimentation rate was 33 mm./1st hr. He attended for antiluetic treatment very irregularly but in February and March, 1938, serological tests were

negative. There was, however, no significant improvement in the chronic penile ulcers (Fig. 6). In addition, there was still some purulent discharge from the urethra. Gonococci were found on microscopic examination, although, according to the patient, he had had no sexual intercourse for several years. Treatment with sulphonamides was prescribed in April, 1938, and all the ulcers healed. On April 19, 1938, the Frei test was strongly positive and the Ito-Reenstierna test negative. On re-examination in December, 1945, he showed extensive scarring of the penis, but this did not interfere with sexual intercourse. All the ulcers remained healed and the erythrocyte sedimentation rate was 3 mm./1st hr, but the serological tests for syphilis were again positive.

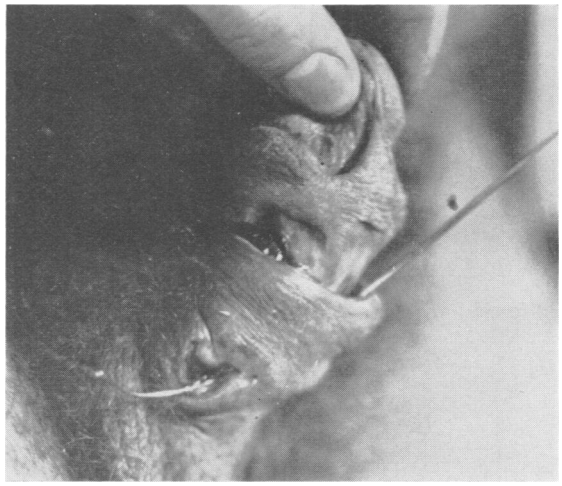

FIG. 6 *Case* 20. 10 *months later* (*March*, 1938). *Extensive ulceration with destruction of subcutaneous tissue and fistula formation*

Case 33, a labourer aged 26, from Helsinki, gave a history of gonorrhoea in November, 1936, but had never had buboes. On June 1, 1937, he developed a small darkfield-negative ulcer (6 × 4 mm.) in the coronal sulcus near the frenulum and a hard, knotty, painless swelling of the prepuce, and 2 days later an enlarged lymph node appeared in the right groin. On June 8 and 10, 1937, the Frei test was positive (with two different antigens). The body temperature was 38·6°C. The erythrocyte sedimentation rate was 46 mm./1st hr. The primary lesion now measured only 2 × 3 mm. The preputial infiltrate on the right side of the penis had grown to the size of a thumb-tip. It extended from the frenulum and the site of the primary lesion in a lateral direction around the right side of the sulcus and prepuce almost up to the dorsal midline, ending in two separately palpable pea-sized infiltrates. The main part of the lateral swelling consisted of an indolent, hard, and irregular conglomerate of infiltrations (Fig. 7). On June 17, the inguinal bubo was 4 × 5 cm. in size, tender, with slight fluctuation. The iliac lymph nodes were also involved, forming a mass the size of a plum. After 5 days the preputial swelling partly

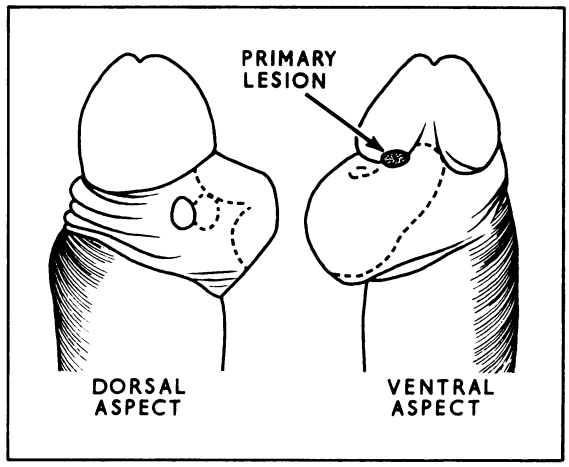

FIG. 7 *Case* 33. *Hard conglomerate of preputial infiltrates*

softened, and the hard infiltrates could be palpated more distinctly as three foci. A lateral infiltrate was connected through a cord formation with the primary site near the frenulum, and from here another infiltrated cord ran parallel with the coronal sulcus to the third focus near the dorsal midline. A draining sinus had developed at the site of the primary lesion and pus could be expressed by compressing the lateral infiltrate. The WR and Kahn test were negative. The erythrocyte sedimentation rate was 56 mm./1st hr. A draining sinus also developed in the most lateral part of the preputial infiltrate.

On June 28, 1937, part of the preputial skin was excised along with the lateral and dorsal infiltrates. Because of its location near the frenulum and glans, the primary lesion was not removed. The wound was closed with catgut and healed by primary intention.

Although the patient had been informed about the serious nature of his disease, he transmitted it to his girl friend, who in July, 1937, developed genital ulcers and bilateral inguinal buboes.

Case 34, a man aged 67, from Helsinki, developed a bubo in the left groin in November, 1937, after intercourse with various women. Examination on December 9 revealed an ulcer, 5 mm. in diameter, in the dorsal coronal sulcus and a bubonulus with central fluctuation in the dorsal mid-shaft. From this bubonulus a 3 to 4 mm. thick lymphangitic cord ran to an infiltrate, the size of a fingertip, at the base of the penis. To the left of the latter another bubonulus was seen in the pubic region. The bubo in the left groin measured 5 × 7 cm. The Frei test was strongly psoitive, the WR and the Kahn test negative. The erythrocyte sedimentation rate was 54 mm./1st hr. On December 11 some pus was aspirated from the bubonulus in the pubic region. The penile bubonulus discharged pus on December 15, after which it was reduced to the size of a pea and the proximal lymphangitis gradually disappeared. A week later, however, a new pear-shaped infiltrate was palpable distal to the bubonulus just above

the prepuce. The erythrocyte sedimentation rate was 103 mm./1st hr. 7 ml. pus were aspirated from the inguinal bubo and proved to be a very good Frei antigen. In January, 1938, the Frei test was strongly positive and the Ito-Reenstierna test negative.

The pear-shaped infiltrate developed fluctuation, and a lymphangitic cord extended distally to the coronal sulcus where pus could easily be expressed from a fistular opening.

In January, 1938, the major part of the penile infiltrate was removed. The wound was closed with catgut only at its upper end, the rest being left open with a drain; it healed within a short time.

The bubonulus in the pubic region continued to discharge pus until May, 1938. In July, 1938, several draining sinuses still remained in the left groin, but they healed rapidly with sulphonamide treatment.

Case 35, a workman aged 36, from Espoo, had intercourse with a promiscuous girl in Helsinki, and 2 to 3 weeks later in December, 1937, a hard lump appeared on the penis, and an inguinal lymph node became tender, reached the size of a plum, and then regressed. He transmitted the disease to his wife, who in January had an ulcer on the vulva and enlargement of the lymph nodes in the left groin. Her Frei test was positive.

When he was first examined on January 18, 1938, there was a preputial infiltrate anteriorly, the size of a thumb-tip, which later developed fluctuation and discharged pus from a draining sinus about 2 cm. above the preputial margin. After 2 weeks another but smaller infiltrate was palpable on the right side of the prepuce (Fig. 8). The WR and Kahn test were negative. The Frei test was strongly positive with two different antigens. The erythrocyte sedimentation rate was 13 mm./1st hr. One month later the lateral infiltrate also became fluctuant and a small amount of pus was aspirated. On March 21, 1938, the preputial sinus was surrounded by a chronic ulcer measuring 25 × 16 mm. (Fig. 9). The infiltration and ulceration increased and perforation through the internal lamina of the prepuce seemed imminent. The affected prepuce was therefore excised, with good results.

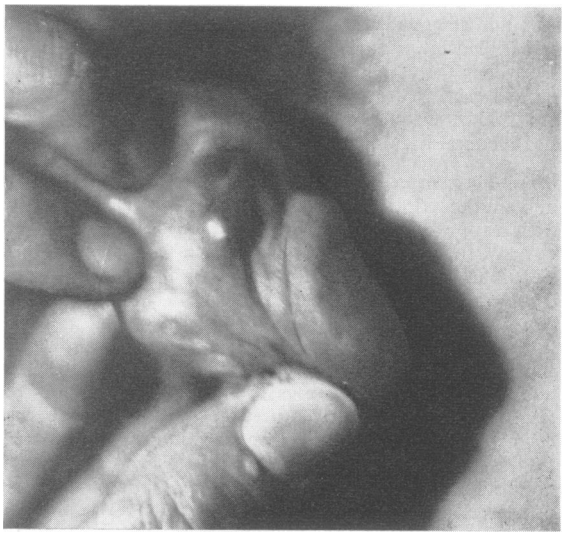

FIG. 8 *Case* 35. *Small bubonulus laterally in internal lamina of prepuce* (*February*, 1938)

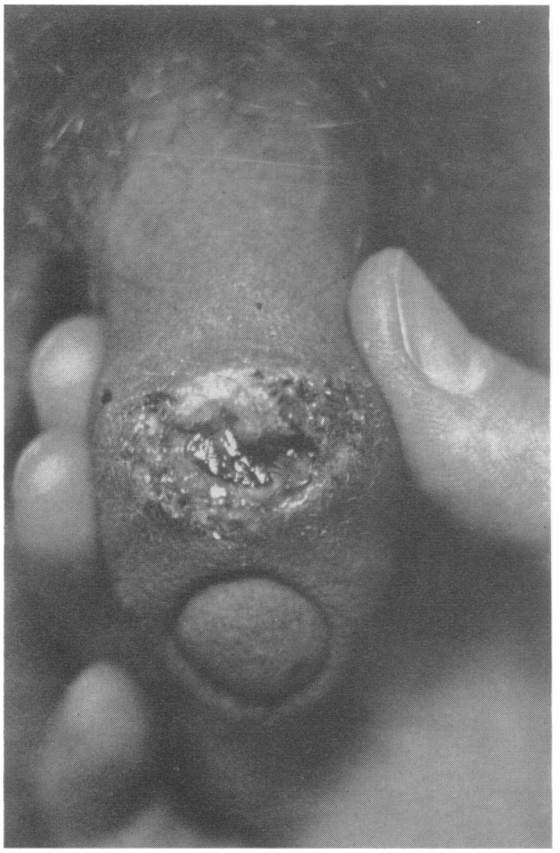

FIG. 9 *Case* 35. 1 *month later* (*March*, 1938). *Preputial sinus surrounded by chronic ulcer*

Discussion

The presence of penile complications in 37 out of 810 patients gives a frequency of roughly 4 per cent. This is the same frequency as reported by Frei (1932) in Germany.

It is not known what factors determine the development of these complications. The age of the patient does not appear to be of importance since penile complications were encountered both in young and old patients; two were under 20, 21 between 20 and 29, eleven between 30 and 39, two between 40 and 49, and one was 67 years old. There were no other accompanying diseases which could be regarded as definite contributory factors.

The rather frequent occurrence of syphilis in patients suffering from lymphogranuloma venereum has been pointed out by several authors. In the Finnish series syphilis was present in 70 (8·6 per cent.) of the 810 male patients (Sonck, 1966). Among 23 females with chronic vulval ulceration there were twelve who had had syphilis. Many of them had been prostitutes, and one had congenital syphilis, so that acquired syphilis was present in about 50 per cent. of these cases (Sonck, 1941). In the present series of 37 males suffering from penile complications, the incidence of syphilis (in fifteen cases) was also comparatively high. In four of these it was probably inactive when the patient contracted lymphogranuloma venereum, but in nearly 30 per cent. (11 out of 37) the two infections occurred in fairly close association.

It is possible that a simultaneous simple balanitic infection could play a role. The influence of trauma must also be considered. The occurrence of chronic vulval ulceration mainly in prostitutes may be due to frequency of intercourse and poor hygiene. Physical activities like horse riding, running, etc., during the acute phase of venereal infections can complicate the course of the disease.

Primary lesions were seen in a very high proportion of cases (30 out of 37). Most of the patients came for treatment at an early stage of the disease so that there was a good opportunity to observe the development of the penile complications. In several cases it was possible to see the primary lesion become smaller and even heal, while active lymphangitis, hard preputial infiltrations, and bubonuli developed in other areas. Later these were seen to rupture and to form ulcers at sites sometimes distant from the primary lesion. Like Cedercreutz (1934a, b) we believe that chronic penile ulcers are often a consequence of lymphangitis causing bubonuli which later ulcerate. This does not mean, however, that the primary lesion could not in some cases progress directly to chronic penile ulceration, as was suggested by Nicolau (1934). However, this appears to us to be less likely. The underlying lymphangitis may not be obvious and it was recorded in only a minority of our cases.

The progressive nature of the penile ulcers is illustrated by the fact that most of the bubonuli resulted in fistulae or sinuses, sometimes involving the urethra, and deformation of the penis could occur. The discharge from the fistulae may remain contagious for several years, as illustrated by one of our patients who transmitted lymphogranuloma to his partner after having had fistulae for over 6 years.

The duration of the lymphogranulomatous process varied markedly from a few months to 7 years. Surgical excision of the affected areas was successful in eight cases, including one with fistulae of 7 years' duration (Case 14). The therapeutic situation was completely changed when sulphonamides were introduced in 1938 and were found to provide an effective treatment for lymphogranuloma venereum. In this series only three patients could benefit from this discovery.

These case histories illustrate the clinical nature of and the sometimes poor prognosis for lymphogranuloma before the advent of the sulphonamides. Cases of this type are hardly ever seen now that satisfactory agents for treating the condition exist.

Summary

The literature regarding the occurrence of infiltrative, ulcerative, and fistular lesions of the penis due to lymphogranuloma venereum is reviewed. Details are given of 37 cases with these complications seen in Finland between 1925 and 1948. Such cases are now rare.

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Lésions infiltrative, ulcérantes et fistulaires de la verge dues à la lymphogranulomatose vénérienne

SOMMAIRE

On passe en revue la littérature sur les lésions infiltrantes dues à la granulomatose vénérienne. On donne des details sur 37 cas présentant ces complications et vues en Finlande entre 1925 et 1948. De tels cas sont maintenant rares.